



CASE REPORT

Huge deep fibrous histiocytoma arising from the sigmoid mesocolon



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KEYWORDS

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Summary Benign fibrous histiocytoma is one of the most common mesenchymal neoplasms of the skin, but it rarely occurs in deep soft tissues. Deep fibrous histiocytoma commonly occurs in the lower limbs or the head and neck region. We report an extremely rare case of deep fibrous histiocytoma arising from the sigmoid mesocolon. A 63-year-old woman initially presented with lower abdominal pain. An image study showed a huge heterogeneous pelvic mass with bleeding. The patient was treated successfully by surgery. We review the literature and discuss the clinical presentation, diagnosis, and management of deep fibrous histiocytoma. Copyright © 2014, Taiwan Surgical Association. Published by Elsevier Taiwan LLC. All rights reserved.

1. Introduction

Benign fibrous histiocytoma (FH) is a tumor composed of a mixture of fibroblastic and histiocytic cells.¹ FH is one of the most common mesenchymal neoplasms of the skin, but rarely occurs in deep soft tissues. Deep benign FH is an uncommon and poorly recognized clinical subtype that arises in subcutaneous or deep soft tissue.² Deep benign FH most commonly occurs in the lower limbs or the head and neck region. We report an extremely rare case of deep benign FH arising from the sigmoid mesocolon.

2. Case Report

A 63-year-old woman, who reached menopause about 7–8 years earlier, presented to our emergency department because of diffuse lower abdominal pain with sudden onset in the morning, associated with mild nausea. According to the patient, she had had frequent attacks of abdominal fullness, especially after meals. However, she denied body weight change, poor appetite, bowel habit change, constipation, or urinary frequency. At our emergency department, vital signs were found to be relatively stable, but physical examination revealed a huge lower abdominal mass with diffuse tenderness and equivocal peritoneal signs. An abdominal computed tomography scan showed a huge heterogeneous pelvic mass with bleeding (Fig. 1). Gynecologic sonography showed mild ascites and one heterogeneous pelvic tumor of about 17 cm × 14 cm × 9 cm; ovarian tumor could not be ruled out.

Conflicts of interest: All authors declare no conflicts of interest.

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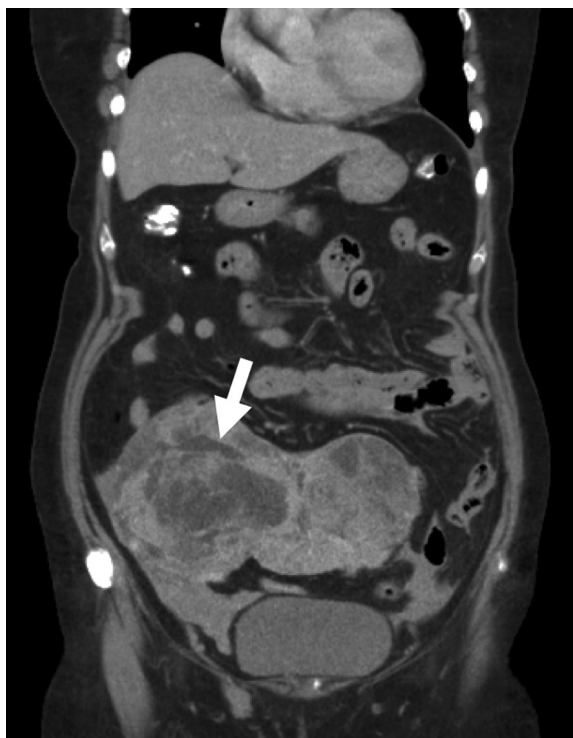


Figure 1 Contrast-enhanced abdominal computed tomography: a heterogeneous tumor, 18 cm × 15 cm × 10 cm in size, arising from the sigmoid mesocolon.

An operation was performed the next morning, and we found tumor rupture with bleeding when the peritoneal cavity was opened. We observed intraoperatively that the tumor was deep-seated and arising from the sigmoid mesocolon. The lesion was totally excised with partial sigmoidectomy (Fig. 2). There was normal atrophied uterus and ovaries. The patient recovered uneventfully and was discharged on the 7th day after the operation.

The surgical specimen was a well-circumscribed neoplasm, which had a storiform growth pattern and



Figure 2 Grossly smooth, well-circumscribed, encapsulated mass lesion, about 20 cm in diameter.

relatively uniform cytomorphology, consisting of spindle-shaped or more ovoid cells with a limited amount of pale cytoplasm. There were focally admixed foamy histiocytes (Fig. 3). Immunohistochemistry and special stains of the tumor cells were diffusely positive for CD34, whereas alpha smooth muscle actin, S-100 protein, keratins, and other special stains were negative. Gastrointestinal stromal tumor, endometrial stromal sarcoma, follicular dendritic cell sarcoma, and melanoma were therefore not likely. The appearance and immunophenotype fitted well with deep benign FH. Based on the surgical and histological findings, deep FH with bleeding was diagnosed. The patient was free of locoregional disease after 12 months of follow-up.

3. Discussion

Benign FH is usually of two types: cutaneous type and that involving deep soft tissues. Histologically, deep FH has many features in common with cutaneous cellular FH, including a storiform and/or short fascicular growth pattern, similar cytologic features with frequently limited or absent polymorphism, and increased cellularity.³ Deep benign FH is an under-recognized neoplasm that most commonly arises in the subcutaneous tissue of the extremities, but may occur at any soft tissue site. The most important diagnostic distinction is the separation of this tumor from the malignant type. Histopathologically, this tumor is a neoplasm of a biphasic cell population of histiocytes and fibroblasts.³ Diagnosis of benign FH is often difficult and usually based on immunohistochemistry. The immunohistochemistry can help in making a differential diagnosis of the soft tissue tumor. Negativity for smooth muscle actin and S-100 can differentiate the lesion from leiomyosarcoma and neurogenic tumors.⁴ Gastrointestinal stromal tumors are one of the most common mesenchymal tumors of the gastrointestinal tract. Most gastrointestinal stromal tumors are c-kit positive (other possible markers include CD34, DOG-1, and desmin). In our case, the immunohistochemistry was not correlated with this criterion. Negativity for CD10, CD56, AE1/3, EMA, calretinin, alpha-inhibin, and D2-40 ruled out gynecologic lesions and

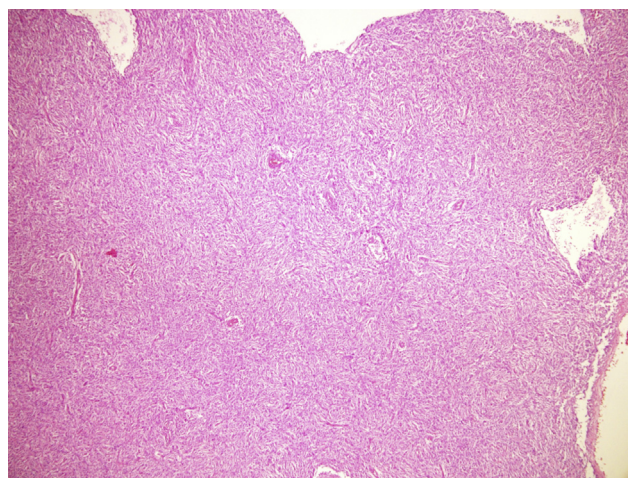


Figure 3 Storiform architecture with staghorn vessels (hematoxylin and eosin, 100×).

that for CD21 eliminated the possibility of follicular dendritic cell sarcoma. MDM2/CDK4 is used for liposarcomas. Malignant FH is composed of malignant pleomorphic sarcomatous cells, bizarre giant cells, and frequent mitotic figures.⁵ The difference between benign and malignant FH is usually obvious, because the latter is a pleomorphic, deep-seated tumor with numerous typical and atypical mitotic figures.⁶

Gleason and Fletcher² reviewed the clinicopathologic features of 69 cases of deep benign FH. The most common anatomic locations were the extremities (58%). Lesions arising in visceral soft tissue were rare. The local recurrence rate of deep benign FH was 22%, which is significantly higher than that of conventional cutaneous FH. Deep FH seems to have an increased risk of local recurrence as compared with conventional FH and also has a very low (and seemingly unpredictable) risk of distant metastasis.

In conclusion, we report a rare case of deep FH arising from the sigmoid mesocolon. To the best of our knowledge, no tumor bleeding case has been reported. Although rare, benign FH must be considered in the differential diagnosis of pelvic tumors. The prognosis of benign FH is excellent if complete excision is performed. Currently, radiation therapy and chemotherapy have no role in the management of benign FH. Our patient had complete excision of the tumor with clear margins, without any morbidity. Regular follow-

up was suggested because of the risk of local recurrence and distant metastasis.

References

1. Ozzello L, Stout AP, Murray MR. Cultural characteristics of malignant histiocytomas and fibrous xanthomas. *Cancer*. 1963; 16:331–344.
2. Gleason BC, Fletcher CD. Deep “benign” fibrous histiocytoma: clinicopathologic analysis of 69 cases of a rare tumor indicating occasional metastatic potential. *Am J Surg Pathol*. 2008;32: 354–362.
3. Calonje E, Mentzel T, Fletcher CD. Cellular benign fibrous histiocytoma. Clinicopathologic analysis of 74 cases of a distinctive variant of cutaneous fibrous histiocytoma with frequent recurrence. *Am J Surg Pathol*. 1994;18:668–676.
4. Fletcher CD, Gustafson P, Rydholm A, Willén H, Akerman M. Clinicopathologic re-evaluation of 100 malignant fibrous histiocytomas: prognostic relevance of subclassification. *J Clin Oncol*. 2001;19:3045–3050.
5. Femiano F, Scully C, Laino G, Battista G. Benign fibrous histiocytoma (BHF) of the cheek: CD 68-KP1 positivity. *Oral Oncol*. 2001;37:673–675.
6. Skoulakis CE, Papadakis CE, Datseris GE, Drivas EI, Kyrmizakis DE, Bizakis JG. Subcutaneous benign fibrous histiocytoma of the cheek. Case report and review of the literature. *Acta Otorhinolaryngol Ital*. 2007;27:90–93.